Imaging Science in Dentistry 2023; 53: 83-9
https://doi.org/10.5624/isd.20220175

A case report of an unusual temporomandibular joint mass: Nodular fasciitis

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ABSTRACT

Nodular fasciitis (NF) is a benign myofibroblastic proliferation that grows very rapidly, mimicking a sarcoma on imaging. It is treated by local excision, and recurrence has been reported in only a few cases, even when excised incompletely. The most prevalent diagnoses of temporomandibular joint (TMJ) masses include synovial chondromatosis, pigmented villonodular synovitis, and sarcomas. Cases of NF in the TMJ are extremely rare, and only 3 cases have been reported to date. Due to its destructive features and rarity, NF has often been misdiagnosed as a more aggressive lesion, which could expose patients to unnecessary and invasive treatment approaches beyond repair. This report presents a case of NF in the TMJ, focusing on various imaging features, along with a literature review aiming to determine the hallmark features of NF in the TMJ and highlight the diagnostic challenges. ***(Imaging Sci Dent 2023; 53: 83-9)***

KEY WORDS: Temporomandibular Joint; Fasciitis; Diagnostic Imaging; Magnetic Resonance Imaging; Tomography, X-ray Computed

Introduction

Nodular fasciitis (NF) is a benign myofibroblastic proliferation typically occurring in the subcutaneous tissue, fascia, and muscle.[1] It was first reported by Konwaler et al.[2] as a subcutaneous pseudosarcomatous fibromatosis. NF presents as a rapidly enlarging mass and patients often complain of soreness and tenderness in the involved region. NF could involve any anatomical site, but the most common locations reported are the upper extremities, the trunk, the lower extremities, and the head and neck area.[3,4]

Recurrence of NF has been reported rarely. Previous studies have reported a recurrence rate of 2%-4%,[5,6] and only a few such cases have been reported.[7,8] Bernstein and Lattes,[3] in their pooled analysis with a large case series, even demonstrated that recurrent NF should not be considered a true NF lesion. The primary treatment option for NF is local surgical excision. Cases of successful treatment with less invasive approaches have also been reported. Hutter et al.[9] reported a case of partially excised lesion that spontaneously disappeared after 2-3 months without any further treatment. Graham et al.[10] reported the successful treatment of NF by intralesional corticosteroid injection after incisional biopsy without any further surgical intervention.

To the authors' knowledge, there have only been 28 cases of intra-articular NF reported, of which only 3 were reported in the temporomandibular joint (TMJ).[11-18] The most prevalent diagnosis of TMJ masses includes synovial chondromatosis (SC), pigmented villonodular synovitis (PVNS), and sarcomas.[19] These lesions have a strong tendency to recur; therefore, an aggressive treatment approach such as excision with a large safety margin is preferred. In cases of advanced lesions, additional systemic therapies such as chemotherapy and/or radiotherapy are applied. Due to its rapid growth and destruction of surrounding structures, NF is often misdiagnosed as a sarcoma, potentially leading to

Received October 6, 2022; Revised October 28, 2022; Accepted November 25, 2022
Published online January 11, 2023
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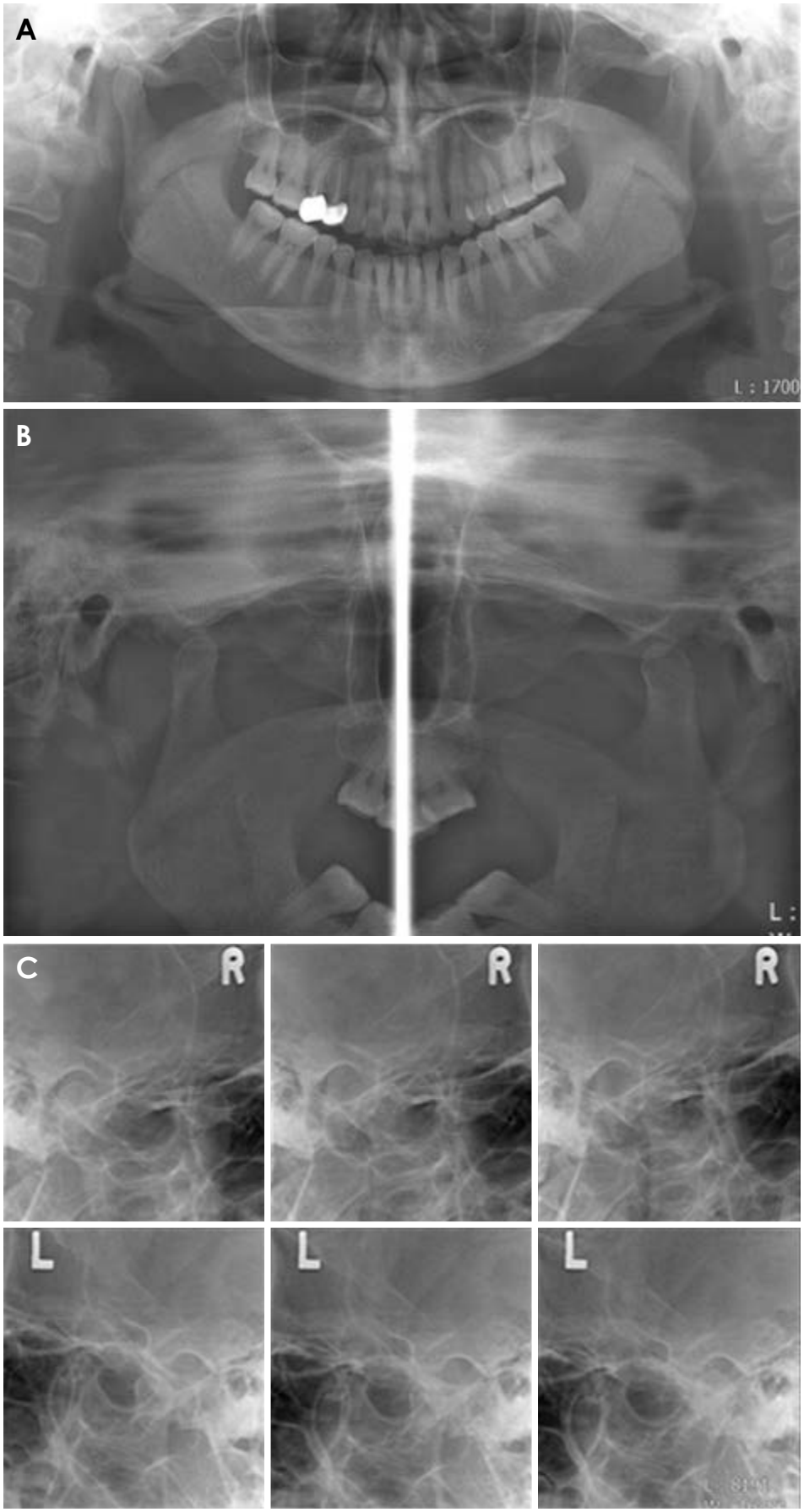

Fig. 1. Panoramic (A), temporomandibular joint panoramic (B), and transcranial (C) radiographs taken at the initial visit show no evidence of pathologic bony changes.

unnecessary and destructive therapy beyond repair.

This report presented a rare case of NF involving the TMJ. The purpose of this report is to highlight the diagnostic challenges, focusing on various imaging features, and review previous reports to determine the hallmark features of the lesion to improve diagnoses in the future.

Case Report

A 54-year-old woman presented to the Department of Oral Medicine at Seoul National University Dental Hospital with a 4-month history of pain during mouth opening in the left posterior auricular area extending to the inferior border of the mandible. She also complained of random episodes of slight hearing loss, but reported no history of trauma. She underwent several sessions of physical therapy in the TMJ region, repeated prescriptions of anti-inflammatory agents, multiple Botox injections, and the implementation of oral appliances at several different clinics. All were ineffective. A clinical examination revealed a slight limitation of mouth opening and left capsule pain at palpation.

A standard panoramic radiograph and TMJ panoramic and transcranial radiographs were taken, but they showed no sign of apparent pathologic changes in either TMJ (Fig. 1). Cone-beam computed tomography (CBCT) scanning was performed 1 month after the initial plain radiographs. A cortical discontinuity on the posterior surface of the left condyle was observed on CBCT images (Fig. 2). The preoperative diagnosis based on clinical examinations was internal derangement of the TMJ, and the patient was prescribed non-steroidal anti-inflammatory drugs for 5 months, with no significant improvement of symptoms.

Seven months after the initial visit, magnetic resonance imaging (MRI) was prescribed for a further evaluation. It showed a well-defined mass extending from the retrodiscal area into the parotid parenchyma. The main part of the lesion was centered around the mandibular condyle head, surrounding it along the joint capsule. The lesion displaced the mandibular condyle antero-inferiorly, causing erosive changes on the posterior surface of the condyle (Figs. 3A-C). On fat-suppressed (FS) T2-weighted imaging (WI), it appeared heterogeneously hyperintense (Fig. 3D). The medial part of the lesion showed higher T2 signal intensity compared to the rest of the lesion. On T1-WI, the lesion appeared isointense to slightly hyperintense compared to the surrounding skeletal muscles (Fig. 3E). The lesion showed partial enhancement on gadolinium (Gd)-enhanced FS T1-WI (Fig. 3F). The medial part of the lesion showed no enhancement on Gd FS T1-WI. A benign tumor of the retrodiscal tissue was first considered. However, erosion of the condylar cortex and extension of the lesion into the surrounding tissues suggested a more aggressive lesion, like a malignant tumor.

The patient was referred to the Department of Oral and Maxillofacial Surgery for an open biopsy. The pathology report described benign myofibroblastic proliferation. However, the patient continued to complain of persistent pain; therefore, contrast-enhanced computed tomography (CECT) was prescribed for a further evaluation. On CECT, the lesion still displayed aggressive features (Figs. 4A and B),

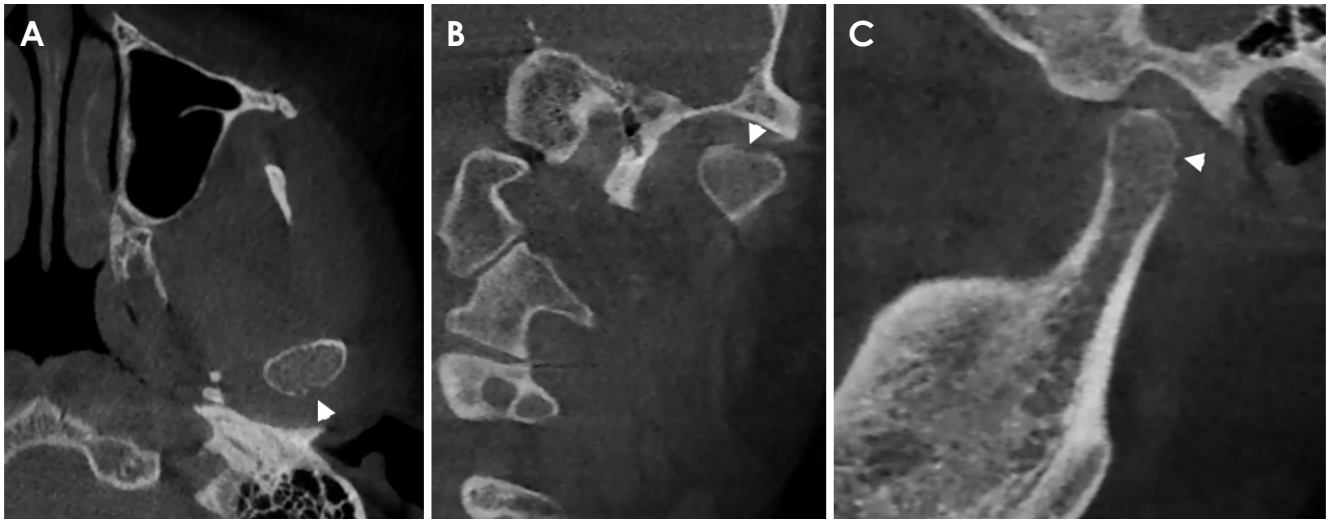

Fig. 2. Axial (A), coronal (B), and sagittal (C) cone-beam computed tomographic images taken 1 month after the initial plain radiographs show a cortical discontinuity (arrow head) on the posterior surface of the left mandibular condyle.

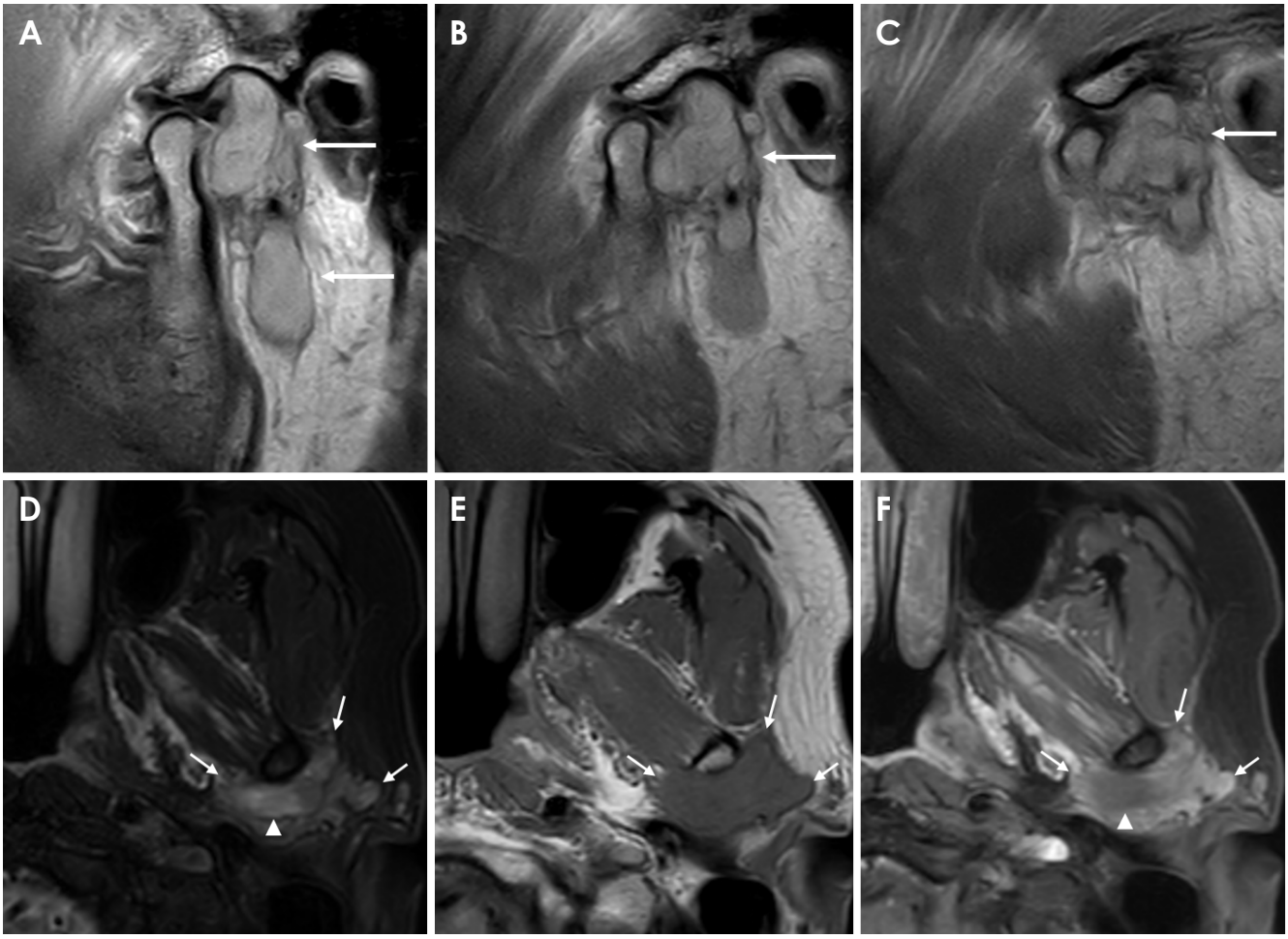

Fig. 3. Magnetic resonance imaging taken 7 months after the initial visit. Serial images of sagittal gadolinium-enhanced T1 weighted images (A-C) show a lesion extending from the retrodiscal area down to the parotid parenchyma. An axial fat-suppressed T2 weighted image (D) shows a hyperintense T2 signal lesion (arrows) with a local increase in signal intensity (arrowhead). An axial T1 weighted image (E) shows an isointense-signal lesion (arrows). An axial fat-suppressed gadolinium-enhanced T1 weighted image (F) shows a heterogeneously enhancing lesion with a medial non-enhancing area (arrowhead).

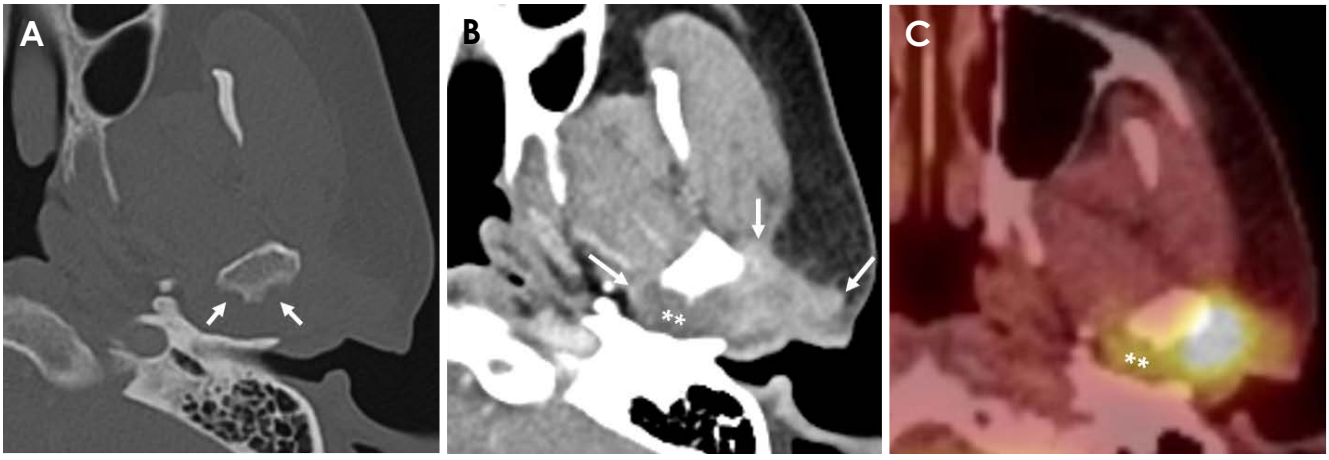

Fig. 4. An axial bone setting computed tomographic image (A), taken 1 month after biopsy shows extensive erosion (arrows) on the posterior surface of the condyle. An axial contrast-enhanced computed tomographic image (B) reveals a heterogeneously enhancing mass with a locally non-enhancing area (asterisk). Positron emission tomography (C) taken 2 weeks later shows a hypermetabolic-signal lesion with less uptake (asterisk) in the medial part.

showing an infiltrative margin, heterogeneous enhancement, and rapid growth. Compared to the previously mentioned MRI, taken about 2 months prior, there was a significant increase in lesion size and cortical erosion on the posterior surface of the left condyle. Moreover, the corresponding lesion displayed a hypermetabolic signal on positron emission tomography (PET) (Fig. 4C). However, the medial part of the lesion, which displayed a hyperintense T2 signal and non-enhancement, also showed no enhancement on CECT and was hypometabolic on PET.

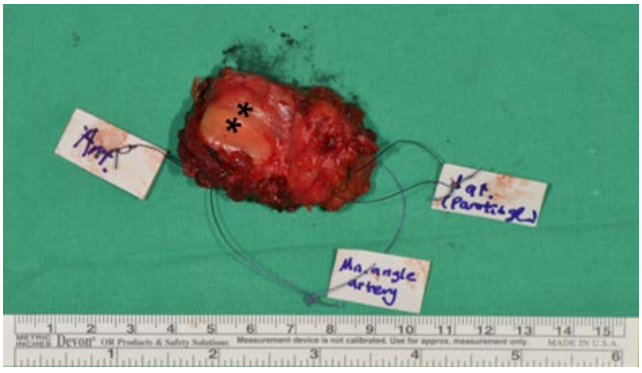

Fig. 5. The lesion is attached to the temporomandibular joint capsule on the superior surface of the condylar head (asterisk), as well as to the postero-lateral surface of the mandibular condyle.

The patient underwent mass resection. The lesion was resected with the left condyle, and the parotid gland was also partially resected. The gross specimen revealed a mass attached to the resected condylar head and the TMJ capsule, confirming capsular origin (Fig. 5).

A histopathological examination of the surgical specimen revealed a well-circumscribed but locally infiltrative mass (Fig. 6A), consisting of bland proliferation of spindle cells, in a myxoid to collagenous stroma. Most areas showed a tissue culture-like growth pattern (Fig. 6B). Extravasated red blood cells (RBCs) and lymphocytes were dispersed throughout the specimen (Fig. 6C). Immunohistochemically, the spindle cells were positive for smooth muscle actin (SMA) (Fig. 6D) but were negative for CD31, with a low Ki-67 labeling index. Taken together, a diagnosis of NF

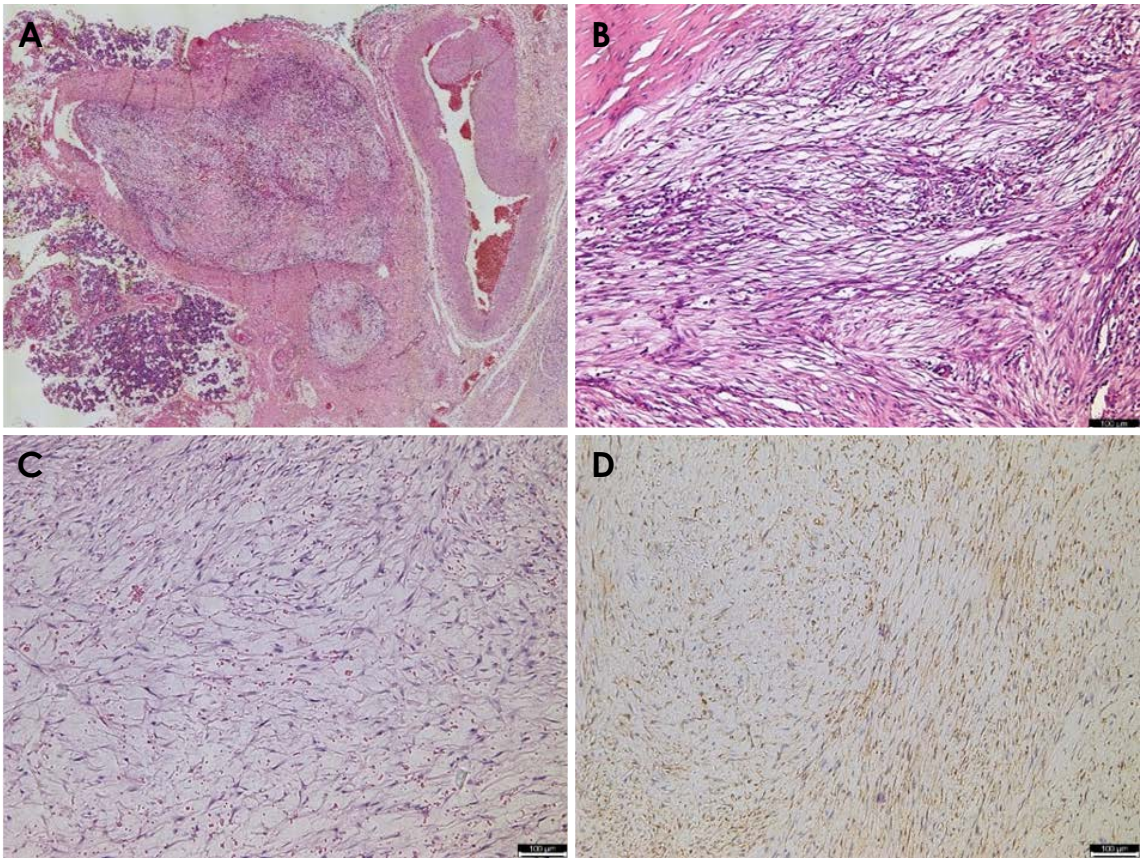

Fig. 6. A. The specimen has a well-circumscribed margin with some areas of local infiltration (hematoxylin and eosin stain, original magnification ×40). B. Bland proliferation of spindle cells, in a myxoid to collagenous stroma (hematoxylin and eosin stain, original magnification ×100). C. Extravasated red blood cells and lymphocytes throughout the specimen (hematoxylin and eosin stain, original magnification ×100). D. The spindle cells are diffusely positive for smooth muscle actin (smooth muscle actin stain, original magnification ×100).

was made. Postoperative follow-up CECT and PET images showed no sign of residual or recurrent lesions.

Discussion

In this case, the mass was located in the retrodiscal area, extending down into the parotid parenchyma. Due to the extent of the lesion, a parotid primary mass extending superiorly into the retrodiscal area needed to be ruled out. A close assessment of the sagittal and coronal view of CECT and MRI indicated that the main part of the lesion was centered around the mandibular condyle head, surrounding it postero-laterally along the joint capsule. The primary diagnosis list was made with retrodiscal tissue or joint capsule-origin lesions. The surgical findings confirmed a mass attached to the condylar head and the TMJ capsule, indicating its capsular origin.

To the authors' knowledge, there have only been 3 reported cases of NF arising in the TMJ.[16-18] Although the exact etiology is unknown, local trauma is considered to be an important etiologic factor, reported in about 10%-15% of cases.[3,7,20] Of the 4 TMJ cases reported, only 1 patient with NF in the TMJ reported a previous history of trauma. This is due to the small number of previous cases; the lack of reports in patients' histories could not be compensated.

In 3 out of 4 cases, the patients complained of pain in the TMJ area. All 4 cases were treated surgically and all reported no recurrence after 9-36 months of follow-up, in accordance with previous reports of NF occurring in other parts of the body.[3] Local surgical excision is the first-line treatment option. However, cases of successful treatment with more conservative approaches, such as steroid injections or partial resection, and even spontaneous regression of lesions, have been reported.[1,9,10]

On the histopathologic examination, NF presents as a well-circumscribed but locally infiltrative, benign proliferation of spindle cells. Extravasation of RBCs was noted, and lymphocytes were widely dispersed throughout the specimen. The NF stained weakly positive on SMA, indicating its myofibroblastic lineage. The specimen was negative for CD31 and low on Ki-67 suggesting its benign nature. Although classified as a benign fibroblastic/myofibroblastic tumor,[21] NF displays very rapid growth and destruction of surrounding structures, mimicking sarcomas on imaging.

Three cases reported CT findings, of which 2 reported erosive changes in the adjacent bony structures. The MRI appearances of NF are reported to be nonspecific and variable. NF is mostly reported as an isointense to slightly hyperintense on T1-weighted images compared to muscle and hyperintense on T2-weighted images. The enhancement pattern can be variable, although in most cases it shows homogeneous enhancement.[22] The 3 cases with MRI findings reported an enhancing mass, with no further descriptions. In the present case, the lesion presented as a well-defined partly enhancing mass with an iso-hyperintense T1 signal and a hyperintense T2 signal on MRI and as a heterogeneously enhancing mass on CECT. Extensive erosion of the adjacent bony structures was evident on both CECT and MRI. The medial part of the lesion displayed distinct features on various imaging modalities compared to the rest of the lesion. It displayed a relatively hypometabolic signal on PET and a high T2 signal with corresponding non-enhancement on both FS Gd T1-WI and CECT. This intralesional change most likely reflected fluid-filled spaces from cystic changes or necrosis, which is consistent with the pathology of NF. This feature has been referred to as an "inverted target" sign and was reported in previous cases investigating MRI findings of NF in other parts of the body.[23,24] This imaging finding can point to a diagnosis of NF, but is not enough for a definitive diagnosis, as central necrotic changes are not uncommon in malignant tumors. Compared with previous reports, we could not find any additional imaging features of NF in the TMJ that could lead to a direct diagnosis. This could be due to the extreme rarity of the lesion, with only 3 other cases for comparison.[4-6]

On the initial visit, the patient complained of hearing loss. However, none of the sectional images taken showed any sign of change in the external auditory canal or along the path of vestibulocochlear nerve. To the authors' knowledge, there have not yet been any reports of the pathognomonic radiographic features of the lesion. A preoperative diagnosis is highly challenging. The rare incidence of NF adds to the likelihood of its misdiagnosis as a more common and/or aggressive entity, such as SC, PVNS and malignant tumors.[7] NF is rarely included on the primary diagnosis list. The treatment of these lesions includes excision with wide surgical margin and systemic therapy, such as chemotherapy and radiotherapy.[23-27] Therefore, it is important to distinguish NF from commonly misdiagnosed, more aggressive lesions, as a misdiagnosis can lead to unnecessary complex treatment. This report provides additional information on the radiological features of these rare lesions to improve the differential diagnosis in the future. Awareness of these unusual entities among oral and maxillofacial radiologists is needed for accurate diagnoses of TMJ masses. At present, the lack of pathognomonic radiographic and clinical features makes a precise diagnosis challenging. Therefore, a

histopathologic examination, such as a preoperative biopsy with ultrasonographic guidance, should precede surgical treatment.

Conflicts of Interest: None

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